

Leiomyosarcoma of Spermatic Cord: Report of One Rare Case and Review of the Literature

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Abstract

A 60-year-old man presented with a 3-year history of painless soybean-like lump in the left hemiscrotum. Physical examination revealed a 2.5 cm tender, nonreducible, firm-to-hard lump palpable over the left spermatic cord. Ultrasonography showed that a well circumscribed 2.4 cm×2.2 cm solid lesion of with scattered blood flow signal was separate from the left testis and epididymis. Intraoperative frozen pathology indicated sarcoma, and thus left transinguinal radical orchidectomy and high ligation of the spermatic cord was performed. Definitive pathological diagnosis of this patient was the leiomyosarcoma of left spermatic cord and negative surgical margins. Due to no positive surgical margins, no lymphatic and distant metastasis, radiotherapy and chemotherapy was not recommended in this case. Regular and close follow-up with thoracoabdominopelvic CT for 68 months shows no signs of local recurrence and distant metastases.

Keywords: Leiomyosarcoma of spermatic cord; Treatment; Prognosis

Case Report

A 60-year-old man presented with a 3-year history of painless soybean-like lump in the left hemiscrotum. In the past two months, the size of the lump suddenly increased and it is always accompanied with pain especially at walking. He has no history of trauma. Physical examination revealed the earthworm-like circuitous expansion of left spermatic vein, and a 2.5 cm tender, nonreducible, firm-to-hard lump was palpable over the left spermatic cord. Ultrasonography showed left varicocele with 2.8 mm inside diameter, and a well-circumscribed 2.4 cm×2.2 cm solid lesion of mixed heterogeneous echogenicity with scattered blood flow signal, and the lump was abutting the upper pole of the left epididymal head, and bilateral testis and epididymis were normal. There were no abnormal findings in blood routine examination, liver and kidney function, LDH, α -fetoprotein (AFP) and β -human chorionic gonadotropin (β -HCG), chest X-ray, abdominal and urinary ultrasonography, abdominal CT. Comprehensive studies did not show any metastases in the lungs, liver, or lymphatic system. An exploratory surgery of left scrotum was performed under subarachnoid block anesthesia on May 27, 2009. Intraoperative frozen section of the lesion indicated sarcoma, and thus left transinguinal radical orchidectomy and high ligation of the spermatic cord was performed.

It was found macroscopically that the lump originated from the spermatic cord near left epididymis, but the lesion had a distinct boundary from testis and epididymis. The lesion was gray and red, solid and fish-like to the touch on the longitudinal cut surface. Microscopic examination revealed that cells arranged in well-defined interlacing fascicles with hyperchromatic pantomorphous nuclei and an abundant amount of eosinophilic cytoplasm, and a lot of vacuoles were found in cytoplasm. There were 3-5 mitoses per 10 high power fields. Small necrotic foci were noted with the appearances of a spindle cell lesion of probable smooth muscle origin. These histological features including high cellularity, pleomorphism, necrosis and mitotic activity suggested it was leiomyosarcoma. The immunohistochemistry revealed positive for Vim, desmin, smooth actin, Ki67 and negative for AE1/AE3, CD117, S-100. Definitive pathological diagnosis of this patient was the leiomyosarcoma of left spermatic cord and negative surgical margins. Patient was discharged a week postoperatively, regular clinical and radiologic follow-up with thoracoabdomino pelvic CT for 68 months shows no signs of local recurrence and distant metastases.

Discussion

The spermatic cord is the most common site of paratesticular neoplasia [1]. The neoplasms of the spermatic cord are mainly benign, including lipoma, fibroma, leiomyoma, hemangioma, cystadenoma, teratoma and dermoid cysts. Malignant neoplasms of the spermatic cord are clinically rare and are generally sarcomas, mainly including liposarcoma, leiomyosarcoma, fibrosarcoma, rhabdomyosarcoma, malignant mesenchymoma, perivascular sarcoma and myxosarcoma. Leiomyosarcoma seems to be the second most common histological variety of adult paratesticular sarcoma following liposarcoma [2]. Though leiomyosarcoma of the spermatic cord is rare, about 120 cases have been reported in the English medical literature so far, and most of the available information derives from small series or case reports [3,4]. Leiomyosarcoma described until now ranges in size from 2-12 cm with a mean of 5 cm [5]. The leiomyosarcoma of spermatic cord occurs mainly in the elderly, with a peak incidence in the 60s and 70s [6]. The leiomyosarcoma of spermatic cord mainly derives from the undifferentiated mesenchymal cells of cremaster muscle and ejaculatory duct [7]. Leiomyosarcoma is subdivided into 3 groups: leiomyosarcoma of the deep soft tissue, leiomyosarcoma of the cutaneous and subcutaneous tissue and leiomyosarcoma of vascular origin. According to the American Joint Committee on Cancer Staging System, paratesticular sarcomas should belong to the deep subtype [8].

Spermatic cord leiomyosarcoma is a rarely encountered malignancy that may be mistakenly diagnosed as inguinal hernia or spermatic cord lipoma. Painless intra-scrotal paratesticular firm mass is the primary symptoms of leiomyosarcoma of spermatic cord. The growth of mass can stop for many years, but suddenly the mass can rapidly grow. Once metastasis occurs, patient's condition fast aggravates, with a poor prognosis. Compared with benign lesion of spermatic cord, there are

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no distinctive imaging findings. Ultrasonography is a useful primary way to assess the mass and its relation to the testis and epididymis, but MRI has more diagnostic values than ultrasonography and CT in revealing mass size, border, infiltration range, and is particularly helpful in better assessing clinical stage [9-11]. With little value in the diagnosis the leiomyosarcoma of spermatic cord, but HCG and AFP have advantages in ruling out testis-derived tumors [12]. Preoperative diagnosis of spermatic cord leiomyosarcoma is difficult and usually made by histological examination. Its behavior is related to the site, histological grade of the lesion and the presence of nodal or distant metastases.

Transinguinal radical orchiectomy and high ligation of the cord is the standard primary surgical procedure, with wide excision of surrounding soft tissue, testis and epididymis. Factors that can affect postoperative recurrence and metastasis include tumor biology, adequacy of surgical resection, and adjuvant treatment. Especially, the importance of negative surgical margins has been well documented [13]. Thus, second wide resection must be performed to remove residual adjacent lesions in patients with positive surgical margin [7]. Even if extensive resection is required to clear positive margins, advances in surgical techniques have made it possible to reconstruct large anatomic defects in this area by local flaps. Local excision followed by additional adjuvant radiotherapy is a suboptimal option. Hematogenous metastasis is the main transfer model of spermatic cord leiomyosarcoma, and the lungs or liver is often affected. Due to less frequency of lymphatic spread, retroperitoneal lymph node dissection (RPLND) is not regularly recommended for patients with spermatic cord leiomyosarcoma patients unless enlarged lymph nodes are encountered on CT scans or palpated during surgery [14]. No report has yet shown a significant survival benefit from the addition of RPLND to radical orchiectomy [15]. Because some cases were accidentally diagnosed by intraoperative or postoperative pathology, few patients always do not receive computerized tomography scan or magnetic resonance imaging, and thus it is difficult to evaluate preoperative radiographic staging. The roles of radiotherapy remain controversial, but most physicians still recommend radical surgery followed by adjuvant radiotherapy to achieve complete tumor regression, and the value of adjuvant radiotherapy has been confirmed in patients with residual lesions [16]. Radiation exposure covers residual lesions, retroperitoneal and ipsilateral pelvic, inguinal lymph nodes and scrotum.

Systemic chemotherapy is rather than radiotherapy to abrogate any possibly hematogenous micrometastases after confirmation of a negative surgical margin [17]. But, conventional adjuvant systemic chemotherapy has no exact efficacy for spermatic cord leiomyosarcoma [18]. At present the role of chemotherapy remains controversial and restricted to the presence of metastatic disease [4]. Up to 75% local recurrence rate in cases treated only using transinguinal orchiectomy has been reported, and the traditional adjuvant therapies are still advisable by most doctors [19]. In our case, histology from the orchiectomy (the specimen) showed negative surgical margins, no lymphatic and distant metastasis, therefore, radiotherapy and chemotherapy was not recommended.

In conclusion, the diagnosis and management of extratesticular lesions can be confusing and troublesome. Spermatic cord leiomyosarcoma, although rare, should be one of the first differential diagnoses for a firm-to-hard lump in the spermatic cord. Intraoperative frozen pathological diagnosis is mandatory for suspected cases, and it is important to evaluate preoperative radiographic staging by

ultrasonography, CT and MRI. Apart from radical orchiectomy with negative margins, efficacy of additional adjuvant retroperitoneal lymph node dissection, radiotherapy and chemotherapy to reduce recurrence is controversial, but each patient must receive intensive long-term follow-up.

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